

The Case of Sacrococcygeal Teratoma in an IVF Pregnancy: Is There any Association between Congenital Tumors and Assisted Reproduction Techniques?

Anjali Tempe¹, Nilanchali Singh^{1*}, Ila Sharma¹, Satish Agarwal²

1- Department of Obstetrics and Gynaecology, Maulana Azad Medical College, New Delhi, India

2- Department of Paediatric Surgery, Maulana Azad Medical College, New Delhi, India

Abstract

Background: Sacrococcygeal teratoma (SCT) is a rare congenital tumor and its association with IVF pregnancy is not clear. There are limited reports of congenital tumors in IVF pregnancy. The exact embryogenesis of SCT is not known but a genetic etiology has been reported. Whether these congenital tumors have any association with assisted reproductive techniques remains obscure.

Case Presentation: In this study, a case of SCT in an IVF pregnancy with donor oocytes was reported. IVF was performed for bilateral tubal blockade and poor ovarian reserve. It was diagnosed antenatally by ultrasonography. Successful surgical treatment was performed in postnatal period and six months follow-up remained uneventful.

Conclusion: The purpose of reporting this case is to emphasize on the possibility of association of congenital tumors with assisted reproductive techniques and hence, the need for screening in these pregnancies. An association could not be detected based on few case reports and therefore, large population based studies are required to elucidate the effect of these reproductive techniques on occurrence of congenital tumors.

Keywords: Assisted, Congenital Tumors, IVF, Reproduction Techniques, Sacrococcygeal Teratoma.

To cite this article: Tempe A, Singh N, Sharma I, Agarwal S. The Case of Sacrococcygeal Teratoma in an IVF Pregnancy: Is There any Association between Congenital Tumors and Assisted. Reproduction Techniques?. J Reprod Infertil. 2014;15(2):109-112.

* Corresponding Author:
Nilanchali Singh, MM-76,
Surya Vihar Colony, Gora-
rampur, Uttar Pradesh,
India, 91-9811343168
E-mail:
nilanchalisingh@gmail.
com

Received: Aug. 29, 2013

Accepted: Dec. 1, 2013

Introduction

In vitro fertilization (IVF) has become a frequent treatment modality in infertile couples and the number of births after IVF conception has a rising trend (1) Risks like multiple gestation and prematurity have definite association with IVF but increased incidence of congenital anomalies in IVF pregnancies has controversial association. There are only few population based studies comparing the risks of congenital anomalies in IVF pregnancies and general population and they found higher incidence in the former, often ranging between 2.4% to 5.8% of births (2-4). The risk of congenital tumors like sacrococ-

cygeal teratoma in IVF pregnancies has not been studied. Sacrococcygeal teratomas are neoplasms originating from pluripotent cells involving all three germ cell layers and occurring in newborns. Their incidence is 1 in 40,000 live births in general population (5). They are usually diagnosed in neonates and children less than 4 years with the female: male ratio of 3:1 to 4:1.6 SCTs are classified morphologically by Altman according to their relative extent inside the body. In type I, the tumor is entirely outside, sometimes attached to the body only by a narrow stalk; in type II, it is mostly outside; in type III, it is mostly inside; and in



Figure 1. Showing lateral view of the baby with Type II Sacrococcygeal teratoma in Prone Jack Knife position. Tumor is marked with solid line and the incision with dotted line

type IV, it is entirely inside (also known as a presacral teratoma or retrorectal teratoma). We could find only one case report of sacrococcygeal teratoma in IVF pregnancy despite the extensive search and we reported it with successful surgical outcome (7).

Case Presentation

A 34 year old patient who was married for eight years presented to us with primary infertility. She had a history of genital Koch's and took a complete course of anti-tubercular treatment two years ago. On complete evaluation, she was found to have bilateral tubal blockage and poor ovarian reserve. We planned to use IVF for her. Endometrial preparation was done by estradiol valerate. After two failed cycles of IVF with her own ova, she conceived during the third cycle of IVF with donor ovum. There was no history of any congenital anomaly or congenital tumors in the father, the ovum donor or their relatives. She received progesterone support and routine antenatal care.

Her regular antenatal check-ups and her normal first trimester scan were normal. First trimester scan and level II scan at 20 weeks of gestation showed normal findings with no congenital anomalies. An ultrasound was done at 32 weeks of gestation and it showed sacrococcygeal teratoma in the fetus, not diagnosed in earlier scans. Apart from this, the antenatal period remained uneventful. Paediatric surgeons consulted with each other and teratoma excision was planned in post-natal period. Betamethasone was given to accelerate lung maturity and Cesarean section was performed at 38 weeks of gestation.

A female baby with birth weight of 2.8 kg was born. Her Apgar score was 9/9 at 1, 5 and 10 min. She had a 10×10 cm swelling in the sacrococcygeal region with a smooth surface and variable consistency. The genitalia or rectum were not involved. The anal sphincter and limb tone were normal. She was operated on the fourth postpartum day via sacral approach. Teratoma was found to be Type II with some intrapelvic extension and was encroaching near the rectal wall which was carefully dissected without any injury. The post-operative period remained uneventful and the wound healed well. The histopathology report was suggestive of mature teratoma. The baby was fine during the six month follow-up.

Discussion

The exact embryogenesis of sacrococcygeal teratoma (SCT) is not clear. It has been suggested that these neoplasms result from the disruption of blastogenesis; the process which otherwise results in conjoined twins. The other pool of thought is that germinal cell neoplasms originate from remnants of the primitive streak which fails to differentiate among mesodermal, ectodermal and endodermal tissues in the embryonic disc (7). As assisted reproductive techniques involve embryologic manipulation, possibility of disruption at various stages may occur, which may act as a trigger to SCT and other congenital tumors.

Anatomically, the tumour protrudes from the space between the anus and the coccyx and is usually covered by the skin. At times, the skin may be denuded over the surface leading to uncontrolled bleeding and death (7). Histopathology may reveal mature and immature elements arising from mesodermal, endodermal and ectodermal cells. Rescorla et al. reported 126 patients with SCT and the histology showed mature teratoma in 69%, immature teratoma in 20%, and endodermal sinus tumor in 11% of patients (8).

SCT can be diagnosed antenatally by ultrasonography. It is located in sacrococcygeal area and the tumor has mixed echogenecity with anechoic cystic areas interspersed in hyperechoic areas which can increase vascularity. It may be associated with polyhydramnios and in rare cases show features of hydrops fetalis. Other associated antepartum complications can be cardiomegaly, high output cardiac failure, placentomegaly, bladder outlet obstruction and hemorrhage within the tumor. Maternal Mirror Syndrome or pseudotoxemia is a severe maternal condition associated with sacrococcygeal

teratoma in fetus (9). The clinical features are similar to preeclampsia like hypertension, vomiting, severe oedema and hyperdynamis cardiovascular state. It occurs when the fetus has undergone hydropic changes and the mother mimics them. Preterm labour, preterm rupture of membranes, dystocia and hemorrhage from the tumor during delivery can also complicate these pregnancies. Termination of pregnancy is indicated only when fetal lung maturity is ensured. Cesarean section should be performed in tumors more than 5 cm in size (10). The treatment for this tumor is surgical excision in postnatal period. It should be performed as early as possible to prevent further complications.

Long term follow-up of children with sacrococcygeal teratoma is necessary as there can be recurrence of tumor and functional impairment in later life. In a study comprising of 126 patients, recurrence of tumor was seen in 11% of cases with mature teratoma during the five year follow-up. The recurrent disease had histopathology of mature teratoma in 18.2% of cases and endodermal sinus tumor in the rest (8). Long term follow-up in 14 patients with SCT was evaluated; fecal incontinence was observed in 14%, urinary incontinence in 36% of patients and lower limb dysfunction in 27% of cases (11). The follow-up should include clinical examination, alpha-feto protein levels and radiological imaging for at least three years. The patients had no recurrence or functional impairment during the six month follow-up.

Most of the studies support the theory that increased incidence of congenital malformations in IVF pregnancies is due to differences in maternal characteristics or pre-existing biological factors, such as genetic mutations, that may underlie a couple's infertility and not be related to any aspect of IVF procedure (4). Some suggest that increased risk of congenital anomalies in IVF is the result of hormone treatments and potentially harmful manipulations of sperm and egg are associated with it (12). SCT has also been reported to have genetic basis. Inherited regional tendency to developmental error affecting the caudal embryonic segments has been postulated as the cause of this genetic basis. Some other congenital tumors like glioblastoma multiforme, congenital neuro-epithelial tumor, gliosarcoma and medulloblastoma have been reported to be in association with IVF pregnancies (13-15). A large retrospective cohort study comprising of around 1500 IVF children and 8500

naturally conceived children revealed that the risk of congenital tumors was twice in the former, though the difference was not significant (16). Such reports are few in number and whether these congenital tumors have any association with assisted reproductive techniques cannot be commented upon. We emphasize on conducting large population based studies to evaluate this association.

Conclusion

In this study, a case of sacrococcygeal teratoma in an IVF pregnancy was reported which is, otherwise, the most common congenital tumor. This could have occurred by chance but, may also have an association with assisted reproduction. Most of the studies emphasize on congenital malformations in IVF pregnancies, but none have focused on congenital tumors. Larger population based studies are required to elucidate the effect of assisted reproductive techniques on occurrence of congenital tumors, which is a very less studied aspect as compared to other birth defects. The IVF protocols and medications need more evaluations, if any such association exists.

Acknowledgement

We thank the parents of the neonate who allowed us to use the pictures for publication. We also acknowledge the presence of paediatric surgery team who helped us in managing this case.

Conflict of Interest

The authors declare no source of support and no conflict of interest.

References

1. Chambers GM, Wang YA, Chapman MG, Hoang VP, Sullivan EA, Abdalla HI, et al. What can we learn from a decade of promoting safe embryo transfer practices? A comparative analysis of policies and outcomes in the UK and Australia, 2001-2010. *Hum Reprod.* 2013;28(6):1679-86.
2. Kallen B, Finnstrom O, Lindam A, Nilsson E, Nygren KG, Otterblad PO. Congenital malformations in infants born after in vitro fertilization in Sweden. *Birth Defects Res A Clin Mol Teratol.* 2010;88(3):137-43.
3. Bonduelle M, Liebaers I, Deketelaere V, Derde MP, Camus M, Devroey P, et al. Neonatal data on a cohort of 2889 infants born after ICSI (1991-1999) and of 2995 infants born after IVF (1983-1999). *Hum Reprod.* 2002;17(3):671-94.

4. Ericson A1, Kallen B. Congenital malformations in infants born after IVF: a population-based study. *Hum Reprod.* 2001;16(3):504-9.
5. Chisholm CA, Heider AL, Kuller JA, von Allmen D, McMahon MJ, Chescheir NC. Prenatal diagnosis and perinatal management of fetal sacrococcygeal teratoma. *Am J Perinatol.* 1999;16(1):47-50.
6. Avery GB, Fletcher MA, McDonald ME. Neonatology: pathophysiology and management of newborn. Philadelphia: Lippincott; 1994. p. 1219-20.
7. Wee WW, Tagore S, Tan JV, Yeo GS. Foetal sacrococcygeal teratoma: extremes in clinical presentation. *Singapore Med J.* 2011;52(6):e118-23.
8. Rescorla FJ, Sawin RS, Coran AG, Dillon PW, Azizkhan RG. Long-term outcome for infants and children with sacrococcygeal teratoma: a report from the Childrens Cancer Group. *J Pediatr Surg.* 1998;33(2):171-6.
9. Villa JC, John V, Verghella V. Large foetal sacrococcygeal teratoma complicated by preterm premature rupture of membranes and maternal preeclampsia. *Rev Colomb Obstet Ginecol.* 2007;58(4):322-27.
10. Chervenak FA, Isaacson G, Touloukian R, Tortora M, Berkowitz RL, Hobbins JC. Diagnosis and management of fetal teratomas. *Obstet Gynecol.* 1985 Nov;66(5):666-71.
11. Manzoni C, Canali R, Narciso A, Nanni L, Pintus C. [Sacrococcygeal teratoma: single center experience and functional long-term follow-up]. *Clin Ter.* 2011;162(2):99-106. Italian.
12. Anthony S, Buitendijk SE, Dorrepaal CA, Lindner K, Braat DD, den Ouden AL. Congenital malformations in 4224 children conceived after IVF. *Hum Reprod.* 2002;17(8):2089-95.
13. Rizk T, Nabbout R, Koussa S, Akatcherian C. Congenital brain tumor in a neonate conceived by in vitro fertilization. *Childs Nerv Syst.* 2000;16(8):501-2.
14. White L, Giri N, Vowels MR, Lancaster PA. Neuroectodermal tumours in children born after assisted conception. *Lancet.* 1990;336(8730):1577.
15. Morof DF, Levine D, Stringer KF, Grable I, Folkerth R. Congenital glioblastoma multiforme: prenatal diagnosis on the basis of sonography and magnetic resonance imaging. *J Ultrasound Med.* 2001;20(12):1369-75.
16. Olson CK, Keppler-Noreuil KM, Romitti PA, Budelier WT, Ryan G, Sparks AE, et al. In vitro fertilization is associated with an increase in major birth defects. *Fertil Steril.* 2005;84(5):1308-15.